

## Mucocoeles, Surgery

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### Abstract

Mucocoeles of the paranasal sinus are benign expanding lesions that primarily occur between the fourth and seventh decades of life. Sinus mucocoeles may develop due to the obstruction of the normal passageway by trauma, inflammation, mass lesions, idiopathic, or iatrogenic causes. The most common site for the evolution of a mucocoele is within the frontal sinuses, followed in frequency by the ethmoidal, maxillary, and sphenoid sinuses. The symptoms depend on the location and size of the mucocoele. The symptoms can be classified as nasal, ophthalmologic, intracranial, and cosmetic. A mucocoele can become infected, forming a mucopyocoele, with a risk of infectious complications including meningitis, orbital cellulitis, and osteomyelitis. Today, the first choice of treatment is marsupialization of the mucocoeles using endoscopic sinus surgery. External approaches to frontal sinus are still used, in erosion of the anterior frontal sinus wall and far lateral located lesions.

### Keywords

Mucocoele; FESS; External Approach

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The paranasal sinus mucocoele is a dilated mucous-filled sinus that is lined by mucous membranes [1]. Mucocoeles of the paranasal sinus are benign, expanding lesions that primarily occur between the fourth and seventh decades of life. The incidence is similar for both genders [2-4]. Paranasal sinus mucocoeles are extremely rare in children and adolescents. Cystic fibrosis is the main etiologic factor [5]. The symptoms depend on the location and size of the mucocoele.

The mechanism underlying mucocoele formation is multifactorial. Impediments to sinus ostium ventilation are thought to be the principal cause. Sinus mucocoeles may develop due to the obstruction of the normal passageway by trauma, inflammation, mass lesions, idiopathic, or iatrogenic causes [6-8] (Figure I). However, the predisposing etiology of primary and secondary nasal mucocoeles seems to be different. For the formation of primary mucocoeles, inflammatory blockages of mucous drainage, secretory duct obstruction, cystic dilation of mucosal glands, and cystic degeneration of polyps were proposed as possible causes. On the other hand, secondary mucocoeles sequestering residual mucosa in the wound and long-term retention of tissue fluid have also been advocated as factors leading to their formation [9-11].

Bockmühl U et al. has evaluated 290 patients with sinus mucocoele according to their etiologies. They reported incidences related to: a previous operation 66% (168 patients), trauma 14% (37 patients), chronic sinusitis 2% (5 patients), tumor 1% (2 patients), and unknown causes 17% (43 patients). When mucocoeles due to a previous operation were further evaluated, it was concluded that 98% were due to external procedures such as Jansen-Ritter and Caldwell-Luc, and 2% were due to endonasal surgery. On average, the periods of time between external surgery and occurrence were as follows: maxillary sinus 15 years, fronto-ethmoid 13 years, ethmoid sinus 10 years, and frontal and sphenoid sinus 8 years [12,13]. It is reported that the use of alloplastic materials in the management of frontal sinus fractures may be a cause of orbital mucopyocele [14].



Figure I. Frontal sinus mucocoele due to nasal polyposis

Mucocoeles grow slowly. As pressure builds due to continuous swelling, the surrounding bony structures begin to thin, contributing to further resorption and remodeling of the bone. Some authors suggest that release of osteolytic factors may contribute to the progressive development of mucocoeles or mucopyoceles. Prostaglandins and collagenases aid in osteolysis and further enhance the expansile nature of the cysts [15-17]. Lund and Milroy proposed that the obstruction to sinus outflow in combination with superimposed infection caused the release of cytokines from lymphocytes and monocytes. The cytokine release stimulates fibroblasts to secrete prostaglandins and collagenases, which in turn could stimulate bone resorption leading to expansion of the mucocoele [18].

Histopathologically, paranasal sinus mucocoeles have features of respiratory mucosa with cyst walls demonstrating single-layered, pseudostratified, ciliated, columnar epithelium [16,18,19]. Although metaplastic changes are rare, chronic cases show evidence of squamous metaplasia. Reactive bone formation is also possible in areas adjacent to cyst epithelium [16]. Mucocoeles show elevated expression of IL-12 which secondarily leads to increased expression of IL-2 and interferon gamma. Subsequently, an increased activation of TH2 lymphocytes hastens the presence of a chronic inflammatory infiltrate [17]. Additional findings include cholesterol granuloma, hemorrhage, fibrosis, and granulation tissue [20]. Central nervous tissue may also be identified in those cases of intracranial herniation [16]. In some cases, ruptured salivary ducts may be identified. The adjacent minor salivary glands often contain a chronic inflammatory cell infiltrate and dilated ducts. Most mucocoeles are self-limiting. They rupture and resolve on their own. However, other lesions are chronic in nature and local surgical excision is necessary. To minimize the risk of recurrence, when the area is excised, the surgeon should remove any adjacent salivary glands that may be supplying the lesion. The specimen should be submitted for pathologic examination to confirm the diagnosis and to rule out the possibility of a salivary gland malignancy. The prognosis is excellent, although occasionally mucocoeles recur, necessitating reexcision, especially if the associated glands had not been removed [19].

Radiographically, the sinus is initially opacified by retained secretions, which displaces all the air content. Pressure erosion of the surrounding bony walls by the mucocoele decalcifies the mucoperiosteal cortical plate and destroys the normal scalloped borders of the sinus (Figure II). With progression of the lesion, the sinus appears more radiolucent radiographically. There may be a zone of reactive osteitis and bone sclerosis around the margins of the mucocoele. In approximately 5% of mucocoeles, peripheral calcification is evident radiographically. This calcification may be large enough to stimulate the growth of an osteoma. With extensive intracranial extension by a frontal mucocoele, a segment of the posterior wall of the frontal sinus may be displaced as a bony fragment deep into the frontal lobe. A view of the optic foramen may be valuable in the diagnosis of a mucocoele of the frontal sinus as well as of the posterior ethmoidal cells. The sphenoid mucocoele presents radiographically as a destructive expanding cystic or mass lesion.

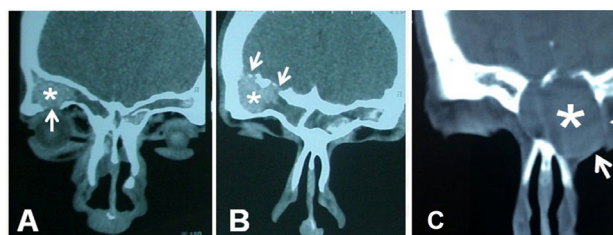


Figure II. The coronal plane CT image showing a mucocoele (asteriks) in the right frontal sinus base that thinned the bone structure (A). In addition, thinning (arrows) and deformation can be observed in the right sinus roof (B) and anterior wall of the left frontal sinus (C).

When expansion and bone destruction are present, differential diagnosis includes benign and malignant lesions of the paranasal sinuses. Benign lesions include neurofibroma; dermoid, epidermoid, cementifying fibroma, angiofibroma, inverting papilloma, and cylindrinoma. Malignant lesions include adenoid cystic carcinoma, plasmocytoma, embryonal rhabdomyosarcoma, lymphoma, schwannoma, and tumors of dental origin [19,21,22]. In the absence of bone erosion, mucocoeles must be differentiated from several conditions including retention cysts, chronic sinusitis, antrochoanal polyp, and polyposis of the paranasal cavities [23-25].

A mucocoele can become infected, forming a mucopyocoele with the risk of infectious complications including meningitis, orbital cellulitis, and osteomyelitis. When the mucocoele expands so that the sinus volume is insufficient to house the mucocoele, pain and pressure erosion can occur [26]. Pressure erosion can result in significant anatomic deformity including facial asymmetry and dynamic proptosis of the orbits [27]. Fortunately, after the resolution of the mucocoele, the deformity usually remodels. Symptoms vary depending on the location of the mucocoele and may be classified as rhinological, neurologic, or most frequently, ophthalmologic [28,29]. The most common site for the evolution of a mucocoele is within the frontal sinuses [25,30]. Sphenoid sinus mucocoele is a rare condition which represents only 1-2% of all paranasal sinus mucocoeles [10]. In a study of 108 patients, Har-El G reported paranasal sinus mucocoele occurrence rates as: frontal and fronto-ethmoid 66 patients (61%), ethmoid sinus 23 patients (22%), sphenoid sinus 12 patients (11%), and maxillary sinus 6 patients (6%) [30]. Caylakli et al. determined that maxillary sinus mucocoeles make up 10% of all mucocoeles [24].

#### Fronto-ethmoid Mucocoeles

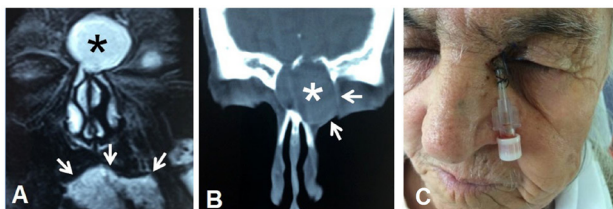
Mucocoeles arising from the frontal sinus present with a variety of clinical signs such as decreased visual acuity, visual field abnormalities, proptosis, ptosis, periorbital swelling, displacement of the globe, restricted ocular movements and choroidal folds. The patient's visual activity is usually not affected unless compression of the optic nerve occurs from the mass effect of the mucocoele. Meningitis, meningoen- cephalitis, pneumocephalus, brain abscess, seizures, and cerebrospinal fluid fistulas are other serious sequelae. The most common presenting symptom was proptosis in 61.5% of the patients reviewed. Forehead swelling and nasal congestion were each present in 38.5% of patients. Other presenting symptoms include orbital swelling, forehead sinus cutaneous fistula, orbital cellulitis, and swelling of medial canthus [25]. Possible initial sequelae include infection, cerebrospinal fluid fistula, meningitis, thrombosis, abscess, paresthesia, and headache. Less com-

mon sequelae include cosmetic abnormality, pyomucocoele, intracranial abscess, headaches, and osteomyelitis. Orbital involvement is rare, but includes vision loss, diplopia, superior ophthalmic vein thrombosis, cavernous sinus thrombosis, and proptosis. Sinus outflow obstruction from trauma, inflammation, previous surgery, tumors, and polyps are exacerbating factors [6]. The definitive treatment of frontal mucocoeles is surgical [32]. Until the 1980's, mucocoeles were exenterated mainly using external approaches. Leading surgical techniques were Lynch-Howarth [33] incision and osteoplastic frontal sinusotomy as described by Bockhmul et al. [34] and by Weber et al. [35]. Traditionally, non-endoscopic treatment of FOE mucocoeles has involved trephination procedures (Figure III) or osteoplastic flap with mucosal extirpation and fat obliteration. In 1989, Kennedy et al. proposed an alternative endoscopic approach that showed no recurrences after a mean followup of 18 months [36]. Today, marsupialization of mucocoeles using endoscopic sinus surgery is considered the first choice of treatment [19,30,32]. It is a safe method with both low morbidity and recurrence rates (Figure IV). External approaches to frontal sinus are still used, alone or combined with ESS, but no clear-cut definition of the limits of a successful endoscopic procedure has been given [32,37,38].

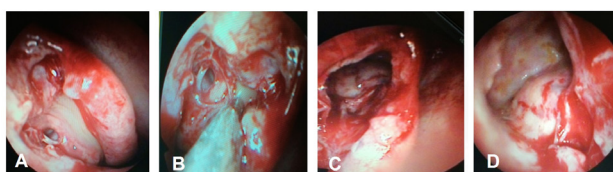
A survey focusing on the current management of frontal sinus disease in the United Kingdom showed that 54% of surgeons are using an external approach to treat frontalsinus mucocoeles independent of their location; only 39% attempt an endoscopic drainage if the cyst lies sufficiently close to the midline [39]. Correa 21 stated that patients with a narrow anterior-posterior diameter of the frontal recess, a highly compartmentalized frontal sinus, an extensive polypoid degeneration of the frontal sinus mucosa, or those with high thickened secretions are candidates for obliteration [40]. Successful frontal sinus obliteration requires meticulous removal of all visible sinus mucosa and the inner cortex of the sinus wall. A wide variety of implant materials for obliteration has been described. Autologous fat transplantation is regarded as the gold standart [41]. The most common reasons for choosing an osteoplastic flap procedure were erosion of the anterior frontal sinus wall requiring forehead reconstruction, far lateral located lesions, or unfavorable frontal recess anatomy with extensive scarring [25]. We choose external approaches in far lateral located lesions, anterior frontal sinus wall's defects, and huge lesions expanding into the intracranial region (Figures V,VI). On the other hand, M. Trimarchi treated frontal sinus mucocoeles with lateral extension by ESS Surgical technique included anterior ethmoidectomy and either Type I, IIa or IIb frontal sinus drainage according to Draf [42,43]. Eloy JA, reported the use of a balloon dilation method for drainage of a large frontal sinus mucocoele [44].

#### Maxillary Sinus Mucocoeles

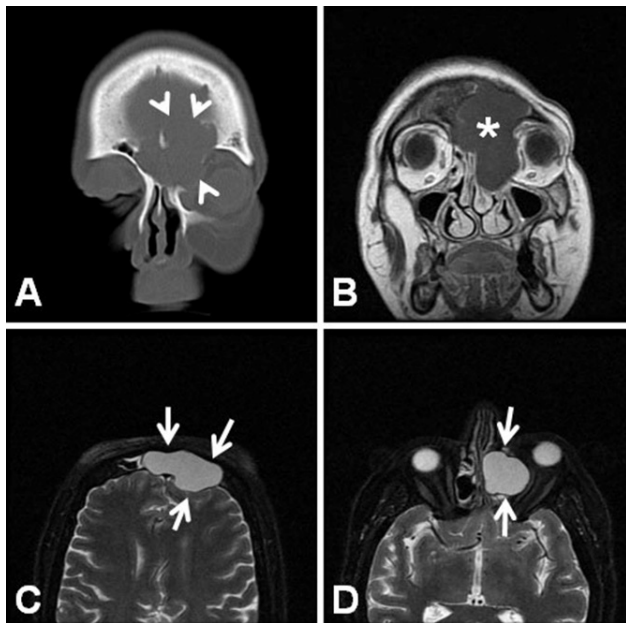
Maxillary sinus mucocoeles are more prevalent in Japan, where they are usually reported as following Caldwell-Luc maxillary sinusotomy [24,45,46]. Mucocoeles that develop following Caldwell-Luc operations are presumed to form as a result of entrapped sinus mucosa. Although one of the theories about development of mucocoele is chronic infection, Busaba et al. compared the bacteriology of maxillary sinus mucocoeles to chronic sinusitis and reported that the data does not support infection as the main origin of nontraumatic maxillary sinus mucocoele [47]. Caylakli et al. state that in 64% of a series of 14 patients, the etiological cause remains uncertain. The symptoms of mucocoeles are related to their expansion and subsequent pressure and the obstruction of surrounding anatomic structures (Figure VII). Antral mucocoeles are commonly reported to present as a painless bulging of the cheek. Medial expansion of the wall of the maxillary sinus into the nasal cavity displaces the inferior turbinate and causes the nasal obstruction [48]. Superior expansion of the antrum into the inferior orbit can cause



**Figure III:** Frontal sinus trephination in a patient that cannot be intubated due to giant hemangioma of the tongue



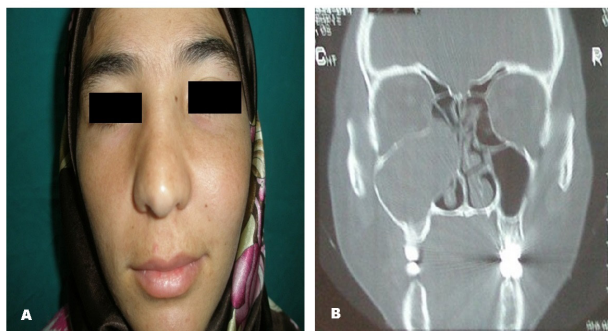
**Figure IV:** Endoscopic marsupialization of fronto-ethmoid mucocoele



**Figure V:** A huge fronto-ethmoidal located mucocoele (asterisk) is shown. In the coronal CT (A) and T1 (B) and T2-weighted MR images (C, D) the expansile mucocoele (arrows) causes thinning (arrowheads) of the frontal sinus walls.



**Figure VI:** The coronal T1-weighted MR image after surgery (E). Air (asterisk) is observed in the left fronto-ethmoidal cavity



**Figure VII:** In the photograph, expansion of the maxillary bone and in CT scan; medial expansion of the wall of the maxillary sinus is seen.

displacement of the orbital contents and visual changes. Downward displacement into the area of the alveolus can even cause a loosening of teeth [46].

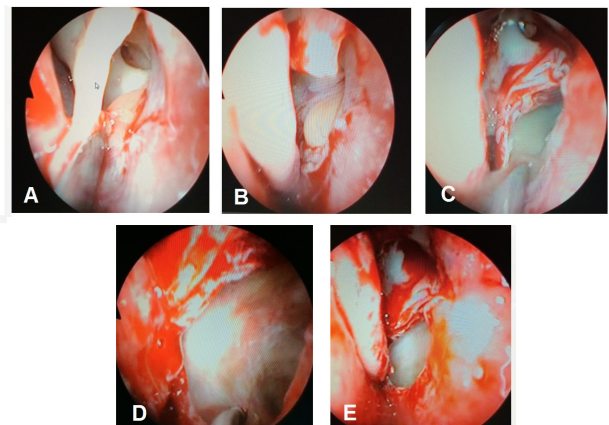
Retention cysts are common in the maxillary sinus and may be found on imaging studies in approximately 9% of the population. They are thought to form due to obstruction of the ducts of seromucous glands in the sinus lining, which results in an epithelium-lined cyst containing mucous or serous fluid. They develop under mucous membranes of the sinus which explains why they are so thin-walled. Radiographically, the cyst is a rounded, dome-shaped, soft tissue mass, most commonly situated on the floor of the maxillary sinus, often containing clear, yellowish fluid. Mucocoeles are associated with obstruction of the duct or natural ostium of any of the paranasal sinuses and grow under the periosteum. Periosteum contributes to construction of the cystic wall, and, as a result, the wall of mucocoele becomes thick and tough. The growing site of the mucocoele is under the periosteum, whereas retention cysts grow under the mucosa of the sinus. This explains why retention cysts are non-expanding, well circumscribed, mucosa-covered masses, whereas mucocoeles exhibit an osteolytic capacity with a tendency to expand along the path of least resistance [24,48,49].

In the treatment of maxillary mucocoeles, surgical techniques including external approaches, marsupialization, Caldwell-Luc procedures, and endoscopy are used [45,46,49]. There was no relapse in a 14-patient series by Caylakli et al. in which endoscopic ethmoidectomy and middle meatal antrostomy were applied. We consider that an extensive marsupialization of the mucocoele using middle meatal antrostomy is sufficient for treatment (Figure VIII).

#### Sphenoid Sinus Mucocoeles

Sphenoid sinus mucocoele is a rare condition which represents only 1-2% of all paranasal sinus mucocoeles [31]. Three pathogenetic mechanisms have been postulated for sphenoid sinus mucocoele: submucosal oedema, secretory duct obstruction, and ostia obstruction. Submucosal oedema may result from allergic rhinitis causing ostia narrowing with obstruction; ostia obstruction may also be due to chronic sinusitis, polyposis, or both [50]. Sphenoid sinus mucocoele formation has been reported as a possible late complication of radiotherapy to the head and neck [51]. Radiation causes scarred mucosa which may occlude the sinus ostium.

Presenting symptoms are non-specific, and patients may be asymptomatic. The clinical symptoms and signs of sphenoid sinus mucocoele are directly related to the sinus anatomy and its contiguous neurological and vascular structures. Important structures are described which are contiguous with the sphenoid sinus: cranial nerves II to VI, cavernous sinus, carotid artery, sphenopalatine artery and nerve, pterygoid canal and nerve, dura, and pituitary gland. Patients with sphenoid



**Figure VIII:** Endoscopic marsupialization of the maxillary sinus mucocoele.

sinus mucocoele become symptomatic when any of these structures are displaced or compressed; mucocoele commonly spreads along the path of least resistance, anteriorly, into the nasopharynx, or anterolaterally, toward the orbit. Sphenoid sinus mucocoele is also more likely to present with visual changes than with inflammatory disease. Visual loss in sphenoid disease has been attributed to optic neuritis or compression [52]. Hejazi et al. reviewed 130 cases of symptomatic sphenoid sinus mucocoele reported up to 2001. The patients presented with these symptoms, in order of decreasing frequency: headache (87%), ocular disorders (85%), amaurosis (58%), oculomotor palsies (55%), nasal symptoms (38%), endocrine disorders (4 of 130) and anhypopituitarism (1 of 130) [53]. Intracranial extension is unusual in sphenoid sinus mucocoele. If extension occurs, it is commonly into the suprasellar region or anterior cranial fossa. Computerized tomography provides valuable information regarding bony erosion, while MRI enables more precise visualisation of the involvement of the optic nerve, dura, pituitary gland, cavernous sinus, and carotid artery [52].

Mucocoeles may show various imaging features, depending on their contents. They often have low attenuation on CT, a low signal on T1-weighted MRI, and a high signal on T2-weighted MRI, due to their high water content. There is usually no enhancement, or at most marginal enhancement, on CT and T1-weighted MRI, whereas many of the lesions in the differential diagnosis show contrast enhancement. Thus, the diagnosis of mucocoele can be suspected pre-operatively and differentiated from craniopharyngioma, pituitary tumor, dermoid, chordoma, epidermoid, aneurysm, metastatic lesions and, in rare cases, an arachnoid cyst. In mucocoeles with a high protein content, T1-weighted MRI shows a lesion which is homogeneously hyperintense centrally with a surrounding rim of hypointense mucosa [53]. Endoscopic drainage is safe and effective in eradication of disease.

#### Middle Concha Bullosa Mucocoeles

A concha bullosa alone is usually asymptomatic. An enlarged concha bullosa may obstruct the middle meatus or infundibulum. Headaches, orbital pain, exophthalmia, nasal discharge, postnasal drip, nasal obstruction, and hyposmia are all possible symptoms of a concha bullosa mucopyocoele. A concha bullosa mucopyocoele may also manifest as migraine headaches [54].

#### Scientific Responsibility Statement

The authors declare that they are responsible for the article's scientific content including study design, data collection, analysis and interpretation, writing, some of the main line, or all of the preparation and scientific review of the contents and approval of the final version of the article.

#### Animal and human rights statement

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. No animal or human studies were carried out by the authors for this article.

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#### Conflict of interest

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